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Case Report

Bone hydatid cyst resulting in fracture: A rare case report

A case of musculoskeletal hydatid cyst

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Abstract

Hydatid cyst (HC) is a parasitic infestation caused by Echinococcus granulosus that can occur in many parts of the body, most commonly in the liver and secondarily in the lungs. Hydatid cyst accounts for approximately 2-3% of all cases. In this study, we aimed to highlight a 32-year-old male patient with an isolated hydatid cyst in the left femur and the radiological findings of HC. The diagnosis of HC was confirmed pathologically.

Keywords

Hydatid Cyst, Echinococcus Granulosus, Musculoskeletal, Albendazole

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Introduction

HC is a parasitic infestation caused by Echinococcus granulosus and humans are an incidental intermediate host for Echinococcus [1]. It is known that most of the infestation affects the liver and lungs [2]. This is because 70% of the cysts drain through the hepatic sinusoids and the rest pass through the lungs[1-3]. Musculoskeletal involvement of hydatid cyst disease may be frequently confused with other lytic/cystic bone lesions. Therefore, it may lead to delayed diagnosis and therefore treatment in cases of musculoskeletal HC. [4]. There is a lack of literature on musculoskeletal hydatid cyst disease suitable for surgical treatment and, to the best of our knowledge, there is no literature on the place of PAIR treatment in bone HC cases. Musculoskeletal HC has been associated with significant morbidity in orthopaedic patients and high recurrence compared to other HC cases [5,6].

The study was carried out in accordance with the provisions of the Declaration of Helsinki and the Good Clinical Practice guidelines. Ethics committee approval was not obtained because the study is a case presentation but oral informed consent was obtained.

Case Report

A 32-year-old male patient presented to the emergency department with a long-standing complaint of pain and swelling in the left thigh and severe pain after a fall. Femur X - ray showed a fracture line in the left femur and a large lytic area in the left femur. The patient with accompanying marked soft tissue swelling was consulted to us for a superficial ultrasound (US) examination with the suspicion of haematoma. US examination revealed a cystic lesion with thick walls and thick septa in the neighbourhood of the left femur [Fig. 1]. Thigh tomography (CT) examination showed a fracture line in the left femoral neck and a lytic area in the proximal left femur [Fig. 2]. Pathological fracture was primarily considered because of the low energy of the trauma and the presence of a large fracture line and large lytic area in the femur with a prominent soft tissue component [Fig. 2]. Magnetic resonance imaging (MRI) of the thigh showed a fracture line with a large lytic area in the left femur and loss of height in the femoral neck [Fig. 3]. It was clearly seen that the lytic area in the left femur was associated with cystic lesions between the neighbouring muscle planes. Multiple cystic lesions with thick walls, some with septa, were observed in the MRI images, the largest of which was 12x5 cm in size in the coronal image [Fig. 3]. This cystic lesion first suggested malignant musculoskeletal cystic lesions. However, in the pathological examination, rock water was found macroscopically and hydatid cyst was diagnosed as a result of Echinococcus granulosus parasites. The patient was hospitalised in the orthopaedic ward and an operation was planned. The patient's fever rose to 38.6 °C. Laboratory tests showed a normal total WBC count of 5.0 x 109 cells/L, eosinophil level of 236 cells/L and a normal erythrocyte sedimentation rate of 15 mm/hour. Liver function tests were within normal limits and hydatid cyst serology was negative. The patient was prepared for elective surgery with consent and antihelminthic treatment was administered for 5 days preoperatively. The patient underwent surgery under general anaesthesia and the adjacent multiple hydatid cyst lesions and the femur infiltrating components of these lesions were tried to be removed and the femur infiltrating parts were debrided. Careful pericystectomy was performed along the surrounding muscle fibres to remove the mass as a block without perforating the cyst wall. Following excision, a thorough cleaning of the surgical field was performed. Multiple drainage catheters were placed in the surgical bed. Postoperatively, the patient was followed up radiologically and clinically once a month for 3 months and then every 3 months.

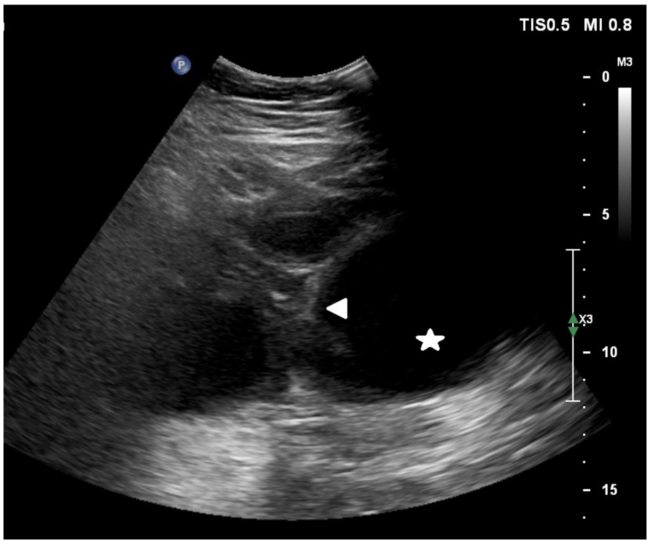

Figure 1. Ultasound examination shows some cystic lesions with thick walls (arrow head) and mostly anechoic internal structure (asteriks)

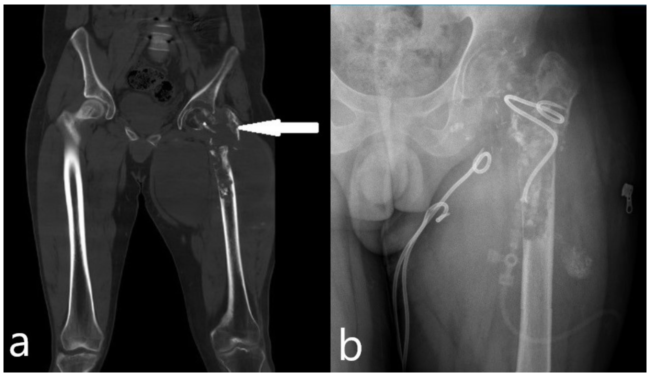

Figure 2. In the non-contrast CT scan, lytic areas and a fracture line are seen on the left femur in the coronal image(arrow). Additionally, soft tissue components in this area are also monitored. b) X-ray image of left femur after treatment.

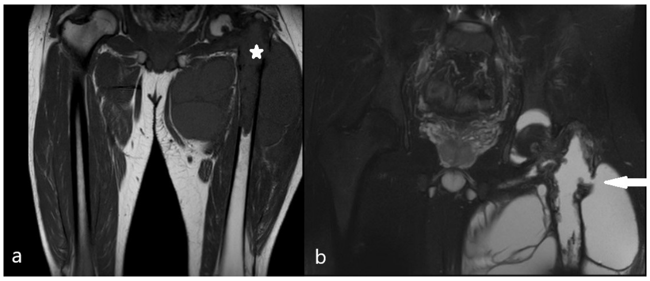

Figure 3. Coronal image T1-weighted (a) and T2-weighted (b) images show thick-walled cystic lesions, some with septa (marked with arrow), that destroy the left femur(marked with asterisk). No significant bone marrow edema or endosteal cupping was observed adjacent to the lytic lesion in the femur

Discussion

The majority of hydatid cysts may occur in the liver, lungs or both, but infiltration into muscle and especially bone is considered an unfavourable site[1, 2]. Although the proportion of primary musculoskeletal HC cases is unknown, musculoskeletal involvement is reported to occur in 0.5% to 4% of HC cases [5, 7]. The most common musculoskeletal sites are pelvic, thigh and paravertebral musculature. Liver and lung involvement is very common due to the route of transmission of HC cases to humans. It is theoretically possible that HC can occur anywhere in the body where the blood circulation reaches, especially in endemic countries such as Turkey[8]. It is widely believed that high lactic acid concentration in skeletal muscle and mechanical factors such as contractions reduce sac formation [1, 6]. Localisation, size and stage of the cyst are most important in forming the clinical picture[4]. Since HC can be seen as simple or complex cysts depending on the stage, it can mimic any cyst [6,8]. Bone HC can mimic early-stage osteomyelitis, and in later stages, the cyst gradually grows and fills the medullary space, as in this case. Bone medulla erosion causes osteolysis and can be radiologically confused with lytic bone lesions such as aneurysmal bone cyst, giant cell tumor, and cystic metastasis. Chronic HC cases in which the bone marrow space is extensively invaded by the parasite may mimic fibrous dysplasia. The main reason that makes the diagnosis of hydatid cyst of bone difficult is that it does not have the typical appearance of classical HC. Pericyst formation does not develop in bone hydatid cyst. Because the pericyst is a fibrotic tissue. Therefore, unlike other HC cases, bone hydatid cysts do not calcify (because the calcified area is the pericyst). However, the soft tissue component of the cyst may calcify. In addition, bone HC cases progress along the bone trabeculae and therefore do not form the classic spherical shape [1]. Especially in complicated cases, as in our case, it may not be recognised. It is very important to formulate a surgical plan according to the HC stage and lesion complications and to prevent the shedding of daughter cysts during surgery [3, 5]. The classical treatment of musculoskeletal HC is surgical excision. However, recurrent cysts have been reported in 1-11% of patients after surgery [7]. Albendazole may reduce the recurrence rate of HC disease and is recommended both pre-op and post-op. PAIR treatment in musculoskeletal cases has been increasing in frequency recently [5, 8]. However, PAIR treatment was not considered in our case due to the fact that our case was both disseminated and infiltrated into the bone.

Conclusion

Although musculoskeletal hydatid cysts are extremely rare, they may cause complications depending on the localisation and stage of the lesion. Diagnosis may be difficult and delays in treatment may occur. This case report is one of the few case reports on aspiration cytology of bone involvement. After the diagnosis, appropriate surgical treatment followed by albendazole treatment minimises recurrences.

Scientific Responsibility Statement

The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and human rights statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or compareable ethical standards.

Conflict of interest

The authors declare that there is no conflict of interest.

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